

Iliac artery endofibrosis

Case study of an elite triathlete

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Progressive stenosis of the iliac artery, known as *iliac artery endofibrosis*, is poorly recognized by primary care physicians. It is mainly found in male endurance cyclists who are younger than 40 years of age.^{1,2} Because vascular disease is unexpected in these athletes, an arterial insufficiency syndrome is often missed; instead, lower limb and sometimes buttocks pain is attributed to musculoskeletal or neurologic causes. Nonatheromatous functional “kinking” along with stenosis or endofibrosis is the main culprit for claudication.³⁻⁷

The condition is almost certainly underdiagnosed in Canada owing to a lack of recognition and no clear unified strategy for the assessment and management of these patients. The delay in diagnosis is profound, averaging between 12 and 41 months, and diagnosis usually occurs only after many months of failed physiotherapy.¹ The failure to diagnose and treat iliac artery endofibrosis in young athletes has career and social implications that could be markedly reduced with better awareness of the condition among family doctors. According to our literature review completed in February 2015, there are no Canadian data published on this high-yield condition.

Case

Our patient was a 25-year-old professional triathlete who had been competing both nationally and internationally for 12 years. He presented initially after having experienced a year of severe abdominal cramps that appeared to be related to the intensity of exercise. The cramps occurred consistently within a few minutes of the transition from biking to running. He was extensively investigated for various potential causes such as gastroesophageal reflux disease, nutritional concerns, and stress and musculoskeletal issues. Findings from his bloodwork, an abdominal ultrasound, an echocardiogram, a treadmill stress test, and upper gastrointestinal endoscopy all showed normal results. Over time, the cramps dissipated but were replaced with left buttock and leg pain. His symptoms, which worsened over the next 6 months, included progressive exercise-induced weakness, cramping, and pain in his left quadriceps, hamstring, gluteus, adductor, and gastrocnemius muscles. These unilateral symptoms were most severe when he ran immediately after a hard effort on the bike, were consistently reproducible if he cycled more than 40 km/hr, and would cease after 2 minutes of rest. His discomfort ultimately resulted in his withdrawal from a world championship competition. He was otherwise healthy, a nonsmoker, and not taking any medications or athletic supplements.

The patient's physical examination, which was performed by his family doctor, did not reveal any abnormalities other than a unilateral grade 1/6 murmur auscultated over the left common iliac artery with flexed hip. The course of investigations included a computed tomography angiogram (CTA), ankle brachial index (ABI) tests (at rest and exertion), and magnetic resonance angiography with flexed and extended hip. His CTA revealed 90% stenosis in the proximal third of the left common iliac artery with poststenotic dilation and collateral flow (**Figures 1 and 2**). Ankle brachial index tests confirmed a tight stenosis but failed to show any decreased waveforms either at rest or with exercise. Magnetic resonance angiography with flexed

EDITOR'S KEY POINTS

- Iliac artery endofibrosis is an underrecognized condition characterized by intimal thickening of the iliac artery. It is mainly found in elite amateur or professional male endurance cyclists who are younger than age 40. As vascular disease is unexpected in these athletes, an arterial insufficiency syndrome is often missed. The condition should be considered early in athletes who experience reproducible claudication symptoms with exercise.
- A focused history, physical examination, and detailed investigations are essential for diagnosis. During physical examinations, listen for iliac artery flow murmurs in flexion and extension. Perform ankle brachial index tests at rest and exertion. Request a computed tomography angiogram and, in appropriate patients, magnetic resonance angiography with flexed and extended hip.
- Failure to diagnose and treat iliac artery endofibrosis in young athletes has career and social implications; a high index of suspicion for this condition is important.



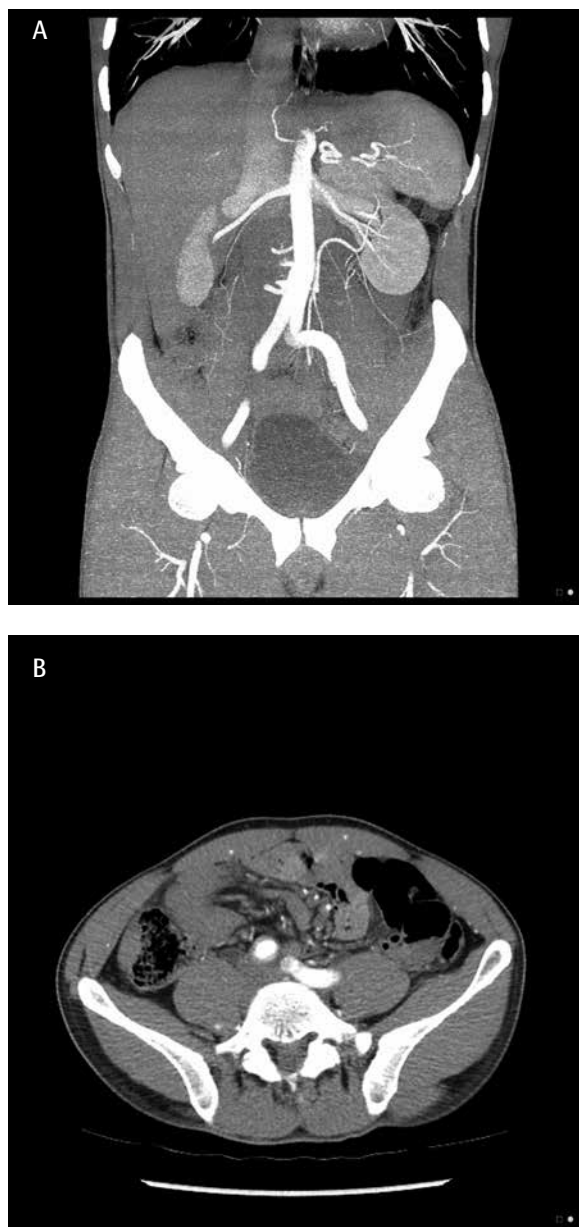
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Figure 1. Computed tomographic angiogram of the abdominal aorta, revealing tight stenosis in the proximal left common iliac artery. The vessel is kinked and is narrowed by more than 90% over a short distance. Poststenotic dilation is noted. The external iliac artery is normal: A) Coronal and B) transverse views.



hip revealed bilateral defects of considerable narrowing of the left common iliac artery, a mild degree of stenosis in the left external iliac artery, and stenosis to a lesser extent in the right external iliac artery. The patient was referred to a vascular surgeon and to a sports medicine physician who had experience

Figure 2. A 3-dimensional reconstruction of the patient's computed tomographic angiogram of the abdominal aorta and common iliacs, revealing 90% proximal stenosis in the left common iliac



in this area. The patient underwent a 3-hour left iliac artery repair with shortening of the artery and a vein patch. He had an uncomplicated postoperative course and returned to his previous level of performance within 8 weeks. He went on to win multiple national championships and competed at the Olympic games. Ultimately, he developed similar ischemic symptoms in his right lower extremity, which placed him in the minority (15%) of athletes who suffer from bilateral iliac artery endofibrosis.⁸ He later underwent uncomplicated surgical repair for the right side as well.

Discussion

The number of clearly identified cases of sports-related endofibrosis is relatively low owing to a lack of awareness, the discrepancy in diagnostic criteria, and the difficulty in establishing a reliable imaging modality.

Published data are limited mainly to case series, isolated case reports, and a single high-profile systematic review.^{1,6} Among the paucity of North American literature on this topic, published Canadian data are lacking.

Those at greatest risk of iliac artery endofibrosis are elite amateur or professional cyclists younger than 40 years of age; it is estimated that 10% to 20% of this population is affected, and 80% of those who are affected are men. At onset of symptoms, these athletes will have

cycled an average of 120 000 km, completing between 14 500 km and 20 000 km per year.⁹


A focused history and physical examination to establish blood flow limitation should successfully identify nearly 80% of cases.⁹ There is no criterion standard for investigation; however, ABI measurements that are less than 0.5 in the supine position completed 1 minute after exercise can identify 80% of cases with 100% specificity for the condition.¹ Laboratory investigations should rule out endothelium-disrupting metabolic disorders such as hyperlipidemia and diabetes. Additionally, conditions that affect blood flow (eg, anemia) should be investigated and treated.

Conservative management involves limiting the time spent in the activity. For those who continue to cycle, positioning adjustments aimed at minimizing hip flexion are recommended, and athletes should be encouraged to avoid pulling upward on the pedals, as this might reduce psoas hypertrophy. However, while acceptable to amateurs, these restrictions are not realistic for professional athletes and this group will likely need surgical correction.¹ Outcomes are generally favourable with minimal complications²; however, owing to the nature of the intervention, surgery should be undertaken only after careful assessment of the inherent risks of vascular surgery. Although it has been suggested that in the long term untreated endofibrosis might predispose patients to atherosclerosis, this has not been established.¹⁰

Our patient did not have a classic presentation. His initial complaint was abdominal pain rather than the typical claudication, which led to some degree of delay in his diagnosis. We believe his atypical presentation could be related to the plausible theory of collateral stealing phenomenon. His exercise-induced abdominal complaints could be attributed to a shunting of blood from the mesenteric system to supply the lower extremity, which would result in abdominal pain during intense exercise. He also presented with 90% stenosis of his left common iliac artery instead of the more typical external iliac artery involvement. He subsequently developed right-sided symptoms; only 15% of patients who suffer from endofibrosis have bilateral disease.⁸

Conclusion

Iliac artery endofibrosis is an underrecognized condition characterized by intimal thickening of the iliac artery. It should be considered early in athletes who experience reproducible claudication symptoms with exercise. A focused history, physical examination, and detailed investigations are essential for diagnosis. During physical examinations, listen for iliac artery flow murmurs in flexion and extension; palpate distal pulses. Perform ABI tests at rest and exertion. Consider CTA and, in appropriate patients, request magnetic resonance angiography with flexed and extended hip. Symptoms of endofibrosis might change over time, and it might be beneficial to repeat tests that previously revealed negative results for flow limitation at exertion.

This case study highlights the importance of maintaining a high index of suspicion for iliac artery endofibrosis. We hope that family physicians will expedite investigation and management of these patients. 

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Competing interests

None declared

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